Metachronous Thyroglossal Duct Cyst and Inferior Parathyroid Cyst: A Case Report

Chueh-Yi Cheng,[1] Yih-Leong Chang,[2] Jong-Kai Hsiao,[3] and Cheng-Ping Wang[1,4]
Departments of [1]Otolaryngology, [2]Pathology, and [3]Medical Imaging, National Taiwan University Hospital and National Taiwan University College of Medicine, Taipei, and [4]Department of Otolaryngology, National Taiwan University Hospital, Yun-Lin Branch, Yun-Lin, Taiwan.

Thyroglossal duct cysts (TDC) are the most common cervical cysts and are usually considered to be a benign embryonic malformation where the thyroglossal duct fails to obliterate. Parathyroid cysts (PTC) are a rare disease and may also result from malformation of the remnants of the third pharyngeal pouch. Although several sporadic cases of concurrent TDC with other head and neck malformations have been reported in the literature, a combination of TDC and PTC has never been reported. Here, we report the first case of a 35-year-old woman with a metachronous TDC and PTC. The embryologic origins of TDCs and inferior PTCs are revealed from the experience of this case.

Key Words: branchial malformation, parathyroid cyst, pharyngeal pouch, thyroglossal duct cyst
(*Kaohsiung J Med Sci* 2008;24:487–91)

Thyroglossal duct cysts (TDC) are one of the most common benign midline cystic neck masses, especially in the pediatric population. TDCs result from a failure of the thyroglossal duct to obliterate after descent of the thyroid gland [1,2]. In clinical practice, most TDCs present alone without other congenital embryologic malformations. Relatively few sporadic cases were reported to have TDCs combined with other malformations in the head and neck regions, such as thyroid hemiagenesis, branchial cleft cyst, epidermoid cyst or dermoid cyst [3–7]. However, a TDC in combination with a parathyroid gland cyst (PTC), which is a rare disease and may result from malformation of the embryologic remnants of the third pharyngeal pouch, has never been reported in the literature. Here, we report a patient with a metachronous combination of a TDC and a PTC, and we try to uncover the association between these malformations.

Case Presentation

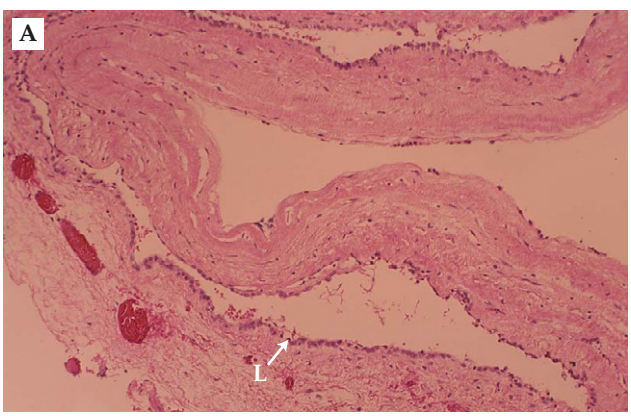
A 35-year-old woman, who denied having any systemic disease, had a history of TDC and presented with an anterior midline neck mass near the hyoid bone, which she has had since she was a child. She underwent a Sistrunk operation for the TDC at the age of 12 and did not have a recurrence until 19 years later when she had another neck mass at the left thyroid region. She had no sore throat, odynophagia, dysphagia, dyspnea or hoarseness. On physical examination, the neck mass was 4 × 3 cm in size and moved upward with the thyroid gland when she swallowed. The neck mass disappeared after repeated fine-needle aspirations, but always recurred 3 months later. Recently, however, the neck mass had enlarged rapidly and persisted after the last aspiration.

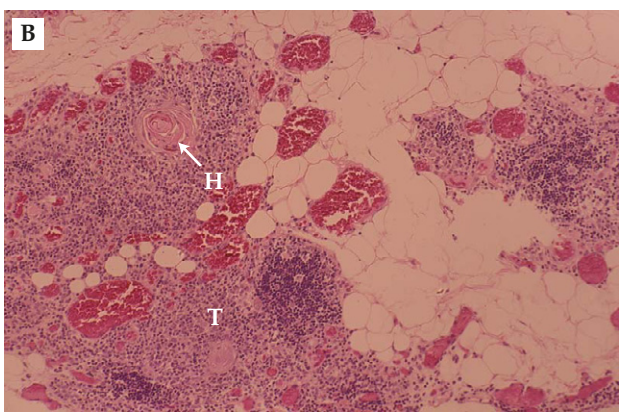
Received: Nov 21, 2007 Accepted: Jan 30, 2008
Address correspondence and reprint requests to: Dr Cheng-Ping Wang, Department of Otolaryngology, National Taiwan University Hospital, 7 Chung-Shan South Road, Taipei 100, Taiwan.
E-mail: wangcp@ntu.edu.tw

Ultrasonography revealed a cystic lesion just posteroinferior to the left thyroid gland. Fine-needle aspiration yielded 18 mL of clear, colorless fluid. Cytologic examination showed few histiocytes and lymphocytes but no malignant cells. Levels of intact parathyroid hormone (iPTH) of the aspiration fluid was markedly elevated (iPTH, 265 pg/mL; reference range, 12–72 pg/mL), although serum calcium level was within the normal range. Technetium-99m thyroid scan revealed a normal thyroid gland without a cold space-occupying lesion. Thyroid function test showed normal results. Computed tomography of the neck showed a 6 × 4 × 3 cm cystic tumor with a very thin cystic wall. The cystic lesion was adjacent to the posteroinferior part of the left lobe of the thyroid gland and extended into the anterior upper mediastinum (Figure 1).

During operation, the smooth, semitransparent thin cyst, which was loosely attached to the thyroid gland, trachea, cervical esophagus, common carotid artery, internal jugular vein and brachiocephalic vein, was easily dissected as a whole from the adjacent structures through a suprasternal transverse cervical approach without resection of the sternum. The left recurrent laryngeal nerve was preserved during dissection. Pathologic examination showed a cystic lesion with a fibrous wall lined by simple cuboidal cells (Figure 2A). Both parathyroid tissue and thymic tissue, including Hassall corpuscles, were found in the cystic wall (Figure 2B), and were confirmed by immunohistochemical stains with cytokeratin and PTH antibodies

Figure 1. *Computed tomography of the neck with contrast enhancement revealed a thin-walled cyst at the anterior mediastinum with lateral and posterior displacement of the trachea, esophagus and greater vessels.*





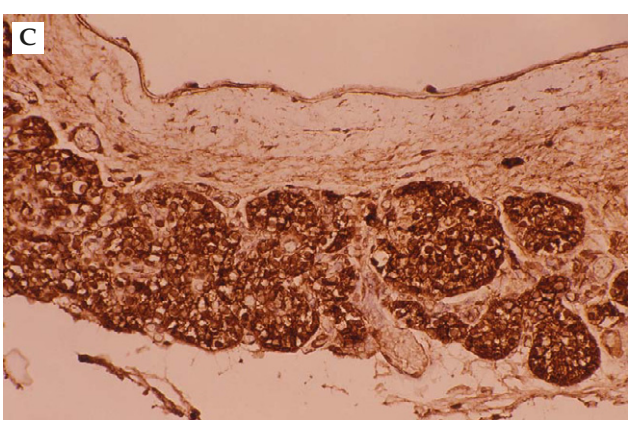


Figure 2. *(A) Pathologic findings revealed a cystic lesion enclosed by a simple cuboidal lining (L) (hematoxylin & eosin, 33×). (B) Thymic tissue (T), including Hassall corpuscles (H) (hematoxylin & eosin, 33×). (C) Parathyroid tissue found in the cystic wall (parathyroid hormone stain, 66×).*

(Figure 2C). After the operation, the patient experienced no complications such as hemorrhage, hypocalcemia, hypercalcemic crisis, tetany and recurrent laryngeal nerve palsy, and did not have recurrence during a 2-year follow-up period.

Discussion

The most important structures of the head and neck are formed by six pairs of branchial/pharyngeal arches. Initially, these arches are separated by deep clefts, known as branchial clefts externally and pharyngeal pouches internally. Each pharyngeal pouch will develop into important structures during later embryologic life. In contrast, the branchial clefts will be obliterated during development. If not obliterated, congenital cervical cysts will occur, such as the second branchial cleft cyst, which is the most common type.

The thyroid gland is originally located in the floor of the pharynx between the tuberculum impar (the first pharyngeal arch) and the copula (the second and third pharyngeal arches) during the 4th week of fetal life [8]. During development, the thyroid gland reaches its final position in front of the trachea and leaves the thyroglossal duct, a narrow canal with an epithelial lining along the descending route of the thyroid gland. Normally, the thyroglossal duct completely disappears before the 10th week [2,8]. However, if the thyroglossal duct is not obliterated, the secretory epithelium of the thyroglossal duct may result in a TDC. Therefore, TDCs are usually considered to be benign embryologic malformations that form during development of the thyroid gland and are the most common congenital midline cervical cysts in children, with peak occurrence before 15 years of age [8], as in the case described here.

Parathyroid cysts (PTC) are a rare disease, with a reported incidence of 0.5% of all parathyroid gland diseases [9]. PTCs typically arise from the inferior thyroid gland with left-sided predominance and are more common in middle-aged women. Confirmatory diagnosis of PTC before operation is important owing to differing surgical procedures for PTCs, thyroid cysts and other malformations. However, PTCs are often clinically mistaken for solitary thyroid cysts if unsuspected [10,11]. In general, it is thought that there is no radiologic differentiation that can distinguish TDCs from PTCs. But, if the single cyst contains thin walls and is mainly located posteroinferior to the thyroid gland with extension downward to the mediastinum, PTC must be considered [10,12]. Fine-needle aspiration may play an important role in the differential diagnosis of PTCs from thyroid cysts or intrathyroid TDCs. The cystic fluid of PTCs is typically clear and colorless and has a high level of c-terminal PTH, as shown in our patient [10,11,13].

The pathogenesis of PTCs is uncertain. Three possible origins of PTCs have been proposed [11]. First, PTCs may arise from cystic degeneration of a preexisting parathyroid adenoma or a hyperplastic gland. These PTCs contain fibrous tissue with a scattered nest of parathyroid cells and are usually functional cysts and present with hyperparathyroidism [11,14]. Second, PTCs are thought to originate from an accumulation or retention of secretions, with a gradual enlargement or coalescence of the microcysts in the parathyroid gland, which is supported by autopsy data showing that microcysts are common and increase with age in 50% of normal parathyroid glands [11]. Third, PTCs may be persistent embryologic remnants [11]. The parathyroid glands originate from the 3rd and the 4th pharyngeal pouches. The ventral side of the 3rd pharyngeal pouch forms the thymus, which descends far into the anterior upper mediastinum, and the dorsal part of the 3rd pharyngeal pouch differentiates into the inferior parathyroid glands, which is pulled by the thymus to the dorsal surface of the lower pole of the thyroid gland. A number of small tubules called the canals of Kürsteiner, the embryonic ducts that connect the thymic and parathyroid primordial of the brachial pouch during development, may persist and eventually result in PTCs [11]. From the pathologic findings of this patient, which revealed concurrent parathyroid, thymic, lymphoid adipose and salivary tissues in the thin wall of this nonfunctional cyst, this case of PTC is consistent with the third theory and may belong to one of the embryologic malformations from the 3rd pharyngeal pouch.

Nearly all patients only have either a TDC or branchial malformation. Few sporadic cases of concomitant TDC with branchial cleft cyst have been reported [7]. However, a combination of a TDC and a malformation of the pharyngeal pouch in the same patient has never been reported in the literature. Our patient is the first case with a metachronous TDC and PTC, both of which may result from embryologic developmental malformations of the branchial/pharyngeal

structures of the head and neck regions. From the experience of this case, the embryologic origins of TDC and inferior PTC appear to be associated.

Considering the treatment history of this patient, surgery remains the treatment of choice for metachronous PTC and TDC. The Sistrunk operation, which includes partial resection of the tongue base and central hyoid bone, is recommended for treatment of TDC. Despite other suggested nonsurgical approaches such as aspiration and injection of sclerosing agents [15], surgical excision of the inferior PTC with careful preservation of the recurrent laryngeal nerve via transcervical approach is the best treatment option for PTC, especially when repeated aspiration is unsuccessful or if a functional cyst is present [9].

References

1. Sprinzl GM, Koebke J, Wimmers-Klick J, et al. Morphology of the human thyroglossal tract: a histologic and macroscopic study in infants and children. *Ann Otol Rhinol Laryngol* 2000;109:1135–9.
2. Shahin A, Burroughs FH, Kirby JP, et al. Thyroglossal duct cyst: a cytopathologic study of 26 cases. *Diagn Cytopathol* 2005;33:365–9.
3. Tsang SK, Maher J. Thyroid hemiagenesis accompanying a thyroglossal duct cyst: a case report. *Clin Nucl Med* 1998;23:229–32.
4. Haar JG, Boulos EJ, Sadeghi MH, et al. Association of a thyroglossal duct cyst and a dermoid cyst in the neck. A case report. *Ann Otol Rhinol Laryngol* 1981;90:181–2.
5. Bhansali SA, Chang CH, Hotaling AJ. Pathologic quiz case 2. Epidermoid cyst and thyroglossal duct (TGD) cyst. *Arch Otolaryngol Head Neck Surg* 1989;115:752–5.
6. Tyson RW, Groff DB. An unusual lateral neck cyst with the combined features of a bronchogenic, thyroglossal, and branchial cleft origin. *Pediatr Pathol* 1993; 13:567–72.
7. Sonnino RE, Spigland N, Laberge JM, et al. Unusual patterns of congenital neck masses in children. *J Pediatr Surg* 1989;24:966–9.
8. Organ GM, Organ CH Jr. Thyroid gland and surgery of the thyroglossal duct: exercise in applied embryology. *World J Surg* 2000;24:886–90.
9. Alvi A, Myssiorek D, Wasserman P. Parathyroid cyst: current diagnostic and management principles. *Head Neck* 1996;18:370–3.
10. Ujiki MB, Nayar R, Sturgeon C, et al. Parathyroid cyst: often mistaken for a thyroid cyst. *World J Surg* 2007;31: 60–4.
11. Ippolito G, Palazzo FF, Sebag F, et al. A single-institution 25-year review of true parathyroid cysts. *Langenbecks Arch Surg* 2006;391:13–8.
12. Jarnagin WR, Clark OH. Mediastinal parathyroid cyst causing persistent hyperparathyroidism: case report and review of the literature. *Surgery* 1998;123:709–11.
13. Silverman JF, Khazanie PG, Norris HT, et al. Parathyroid hormone (PTH) assay of parathyroid cysts examined by fine-needle aspiration biopsy. *Am J Clin Pathol* 1986;86:776–80.
14. Fortson JK, Patel VG, Henderson VJ. Parathyroid cysts: a case report and review of the literature. *Laryngoscope* 2001;111:1726–8.
15. Akel M, Salti I, Azar ST. Successful treatment of parathyroid cyst using ethanol sclerotherapy. *Am J Med Sci* 1999;317:50–2.

異時性甲狀舌骨囊腫及下副甲狀腺囊腫 — 病例報告

鄭爵儀[1] 張逸良[2] 蕭仲凱[3] 王成平[1,4]

台灣大學醫學院附設醫院 [1]耳鼻喉部 [2]病理部 [3]影像醫學部

[4]台灣大學醫學院附設醫院 雲林分院 耳鼻喉部

甲狀舌骨囊腫是最常見的頸部囊腫，一般認為是胚胎發育過程中，甲狀舌管沒有關閉造成的良性胚胎畸型；副甲狀腺囊腫則是一種罕見疾病，可能起因於胚胎發育時第三咽囊的殘存物。過去僅有少數病例報告發表同時合併甲狀舌骨囊腫與其他頭頸部畸形之案例，但合併甲狀舌骨囊腫及副甲狀腺囊腫之病例卻未曾被報告過；在本文中，我們報導一例三十五歲女性患有異時性合併甲狀舌骨囊腫及副甲狀腺囊腫。並從這個病例中來討論甲狀舌骨囊腫及副甲狀腺囊腫之胚胎起源。

關鍵詞：腮裂畸形，副甲狀腺囊腫，咽囊，甲狀舌骨囊腫

(高雄醫誌 2008;24:487–91)

收文日期：**96** 年 **11** 月 **21** 日

接受刊載：**97** 年 **1** 月 **30** 日

通訊作者：王成平醫師

台灣大學醫學院附設醫院耳鼻喉部

台北市 **100** 中山南路 **7** 號